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Case report

Stenotrophomonas maltophilia: An uncommon cause of liver abscess

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ABSTRACT

We report an uncommon case of pyogenic liver abscess due to *Stenotrophomonas maltophilia* in an apparently immune-competent individual, the timely recognition of which could avoid a potentially fatal course of infection in the patient. A 45-year-old man, with history of moderate alcohol intake since the last 10 years, was admitted with intense right-sided upper abdominal pain and right-sided chest pain of 10-days duration. Culture of the ultrasound-guided liver aspirate sample yielded a pure growth of *S. maltophilia* identified by the VITEK-2™ automated microbial identification system. Treatment with parenteral levofloxacin and oral trimethoprim-sulfamethoxazole along with pigtail catheter drainage and other appropriate supportive management led to resolution of the abscess with no recurrence of infection at two months follow-up. Physicians need to be aware that *S. maltophilia* infections may not be restricted to hospitalized patients as a low-virulence opportunistic pathogen, but may occur as an important emerging pathogen in community-acquired infections as well.

Introduction

Pyogenic liver abscess (PLA) is a condition of suppurative infection of the liver parenchyma due to bacterial cause. The incidence of PLA has been reported to be 0.5 %–0.8 % in the Western world with a frequency of 20 per 100,000 admissions in hospitalized patients [1]. Although uncommon, it can prove to be fatal if not treated promptly. Common predisposing factors include underlying biliary tract diseases (cholelithiasis, cholecystitis, and malignancies), various biliary procedures such as stenting and sphincterotomy, surgical procedures on the hepatobiliary system, biliary strictures, inflammatory bowel disease, appendicitis, diverticulitis, intra-abdominal infections, abdominal trauma, and cirrhosis [2,3]. Risk factors include diabetes, malignancy, hypertension, alcohol abuse and cardiovascular disease [2,3]. Nearly 55 % of patients with PLA have no clear risk factors and these cases are called cryptogenic [4]. A wide range of microbial agents have been isolated from PLA cases, with either monomicrobial or polymicrobial etiology; the latter being more common [2–5]. Both aerobes and facultative anaerobes like *Escherichia coli, Klebsiella pneumoniae, Pseudomonas aeruginosa, Acinetobacter* spp, *Staphylococcus* spp, *Enterococcus* spp, *Citrobacter* spp, *Streptococcus* spp as well as obligate anaerobes like *Bacillus fragilis*, *Bacteroides* species, *Fusobacterium*, and *Clostridia* have been implicated [2–5]. Stenotrophomonas maltophilia, an environmental Gram-negative bacterium with low virulence, heralded widely as an emerging global opportunistic pathogen, has only rarely been isolated from PLA cases before [6–11].

Here, we report a case of PLA due to *Stenotrophomonas maltophilia* in an apparently immune-competent individual, the timely recognition of which helped in the institution of appropriate therapy and could avoid a potentially fatal course of infection in the patient.

Case

A 45-year-old male patient, truck driver by occupation, was admitted to our hospital with intense right-sided upper abdominal pain and right-sided chest pain of 10-days duration. He also complained of accompanying fever of the same duration. Prior to his admission, he had consulted another hospital and had received some non-specific medication without any relief, for which he was referred to our hospital. The patient gave a history of alcohol intake (moderate amount) since the last 10 years; however, he was otherwise apparently healthy prior to this episode of current illness. He was not diabetic or hypertensive and there were no other significant medical or family history, including history of contact with tuberculosis. On physical examination, the patient was conscious and oriented. He was febrile and recorded a temperature of 37.8 °C. His pulse rate, respiratory rate, blood

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pressure and oxygen saturation were recorded to be 109/min, 22/min, 130/90 mm of Hg and 94 % respectively. On abdominal examination, tenderness was elicited in the right hypochondriac region. Respiratory system examination revealed a right-sided pleural effusion with reduced air entry in right lung and basal crepitations in left lung. Rest of the systemic examination was normal.

Laboratory investigations revealed a raised alkaline phosphatase level of 363 IU/L, a slightly raised total bilirubin level of 1.6 mg/dL (raised direct bilirubin 0.76 mg/dL, normal indirect bilirubin 0.8 mg/dL), and a low albumin level of 2.5 g/dL. Hemogram reports showed an extremely high total leucocyte count of 38.09×10^9/L, neutrophilic leukocytosis with shift to left (polymorphs 90.4 %) and a low hemoglobin level of 117 g/L. Other parameters such as, total red blood cell count (4.85×10^{12}/L) and platelet count (545×10^9/L), respectively were within normal limits. Serum electrolytes, random blood sugar level, and lipid profile were also within normal limits. The patient tested negative for anti-HIV-1/2 antibodies, anti-Hepatitis C virus antibodies, and for Hepatitis B surface antigen. An ultrasonogram of the abdomen revealed a well-defined hypoechoic mass of size $10.7 \times 8.5 \times 7.2$ cm in the right lobe of liver suggestive of an abscess and confirmed by an abdominal computed tomography scan. Subsequently, pigtail catheterization was carried out under local anesthesia which drained approximately 350 mL of thick purulent brownish material which was submitted for various microbiological investigations. The patient was started on parenteral antibiotics (piperacillin-tazobactam and metronidazole) pending culture results. Other supportive management such as maintenance of oxygen saturation, correction of anemia and drainage of pleural fluid was provided as per protocol.

A Gram-stained smear of the abscess material showed plenty of polymorphs (50–70 per low power field) with lots of gram negative bacilli scattered in arrangement (Fig. 1A). Culture on MacConkey agar after 24 h of incubation yielded pure growth of non-lactose-fermenting colonies (Fig. 1B) consisting of oxidase-negative, motile, gram-negative rods identified as *Stenotrophomonas maltophilia* by the VITEK-2^{TM} automated microbial identification system (bioMérieux, Marcy l'Etoile, France). Antibiotic susceptibility testing of the isolate by the standard disc-diffusion test performed as per the Clinical and Laboratory Standards Institute guidelines showed the isolate to be susceptible to chloramphenicol, levofloxacin, minocycline and trimethoprim-sulphamethoxazole, but resistant to ceftazidime and ticarcillin-clavulanate [12]. The individual minimum inhibitory concentrations of the various antibiotics by EzyMIC (HiMedia, Mumbai, India) were as follows - chloramphenicol (1 μg/mL), levofloxacin (1 μg/mL), minocycline (0.75 μg/mL), trimethoprim-sulphamethoxazole (0.25 μg/mL), ceftazidime (≥256 μg/mL) and ticarcillin-clavulanate (32 μg/mL). Other investigations including modified Ziehl–Neelsen stain for acid-fast bacteria, cartridge based – nucleic acid amplification test for *Mycobacterium tuberculosis*, wet-mount for motile trophozoites and culture for fungi were non-contributory. Culture of accompanying pleural fluid specimen as well as blood culture did not reveal any growth.

Antibiotics were changed to a combination of levofloxacin (750 mg once a day intravenous) with trimethoprim-sulfamethoxazole (160 mg + 800 mg two times a day orally) administered for 3 weeks. Simultaneously, drainage with pigtail catheterization was also continued for 2 weeks. Significant clinical and radiological improvement was noted after one week of therapy, with decrease of abdominal pain, remission of fever and significant resolution of abscess and pleural effusion. The laboratory findings also improved gradually. The patient was discharged on the 22nd day of admission, with advice to continue oral trimethoprim-sulfamethoxazole in the same dosage for a further duration of 2 weeks. Clinical examination at 2 months follow-up revealed complete resolution of symptoms without any fresh complaints or any recurrence of infection.

Discussion

Stenotrophomonas maltophilia is an aerobic, non-fermentative, motile, Gram-negative bacillus which belongs to family Xanthomonadaceae [13]. It was first isolated in 1943 from pleural effusion and initially named *Bacterium booker* [14,15]. Subsequently, it was reclassified as a member of the genus *Pseudomonas* and named *Pseudomonas maltophilia* in 1961, later, rRNA cistron analysis led to naming it as *Xanthomonas maltophilia* in 1983 following which, rRNA sequencing led to creating new genus *Stenotrophomonas* in 1993 and appropriately classifying it as *Stenotrophomonas maltophilia* [13–15].

Among the non-fermenters, *S. maltophilia* has emerged as an opportunistic pathogen in immunocompromised patients, with an increase in prevalence from 0.8 to 1.4% during 1997–2003 to 1.3–1.68 % during 2007–2012 along with an estimated attributable mortality rate of 37.5 % [13,15,16]. Due to its low virulence property, it is usually considered an uncommon pathogen in immune-competent individuals [13,16]. However, in recent times infections due to *Stenotrophomonas* has been reported in community-acquired cases in immune-competent individuals as well [17]. Respiratory tract infections (pneumonia) and bloodstream infections (bacteremia) are the most common clinical manifestations of *S. maltophilia* infection. Less common manifestations are urinary tract infections, biliary tract infections, bone and joint infections, skin and soft tissue infections, peritonitis, wound infection, endopthalmitis, meningitis, and endocarditis [13,15,17,18]. Cases of liver abscess infection due to *S. maltophilia* have been very rare. An extensive search of the PUBMED from 1966 for "liver abscess/ hepatic abscess and" AND "*Pseudodomonas maltophilia*/ *Xanthomonas maltophilia*/ *Stenotrophomonas maltophilia*", revealed very

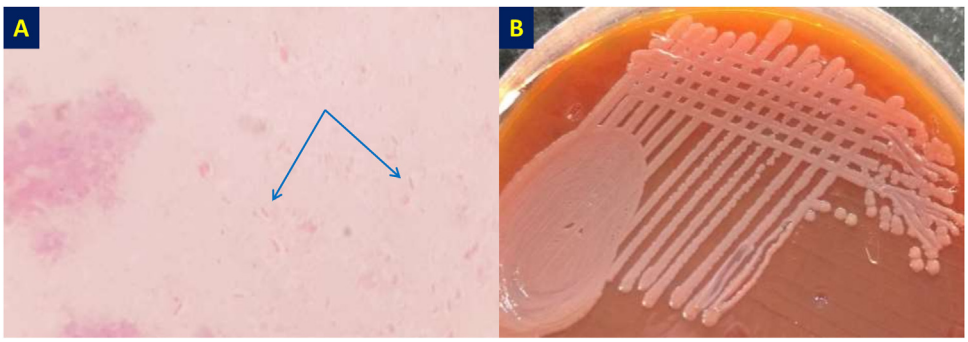

Fig. 1. Liver aspirate showing (A) gram-negative bacilli (1000 ×) and (B) growth of *S. maltophilia* on MacConkey agar plate.

few number of such cases [6–11]. Of these, three cases were in immunocompromised patients comprising of diabetes, human immunodeficiency virus (HIV) infected, and a neonate each [6–8], while, two cases occurred in apparently immune-competent persons [9,10]. One case was described as a part of microbiological profile of pyogenic liver abscesses in patients <70 years age [11]. The outcome was favorable in each of the cases described as single case-reports with complete resolution of infection in all [7–10].

Predisposing factors/comorbid conditions pertaining to *S. maltophilia* infection are well known and include indwelling catheters (central venous catheters and urinary or biliary catheters), mechanical ventilation, recent surgery, malignancies (especially hematological malignancy), intensive care unit admission, organ transplantation, corticosteroids or immunosuppressive drugs, HIV infection, neutropenia, and prior antibiotic use [13,19]. The formation of a liver abscess due to *S. maltophilia* infection, especially in immune-competent persons as in the present case, is thus, a very rare clinical manifestation. In community- acquired infections, items such as sink drains, faucets, water, and sponges, etc., have been identified as environmental sources of *S. maltophilia* in the homes of colonized and noncolonized cystic fibrosis (CF) patients and are considered particularly significant for CF or immunocompromised patients [13]. In the present case, the exact source of the organism remains elusive and the patient might have acquired the infection from any of these environmental sources, especially, in view of his occupation which likely involves a lot of travel.

As regards susceptibility to antimicrobial agents, *S. maltophilia* is known to be intrinsically resistant to a wide variety of antimicrobial agents including ampicillin, amoxicillin, piperacillin, ticarcillin, ampicillin-sulbactam, amoxicillin-clavulanic acid, piperacillin-tazobactam, cefotaxime, ceftriaxone, carbapenems, aminoglycosides and trimethoprim [12,15]. The preferred treatment of *S. maltophilia* infections has been the use of the bacteriostatic compound trimethoprim-sulfamethoxazole [13,15]. Other agents such as the tetracyclines, doxycycline and minocycline, fluoroquinolones, and ticarcillin-clavulanic acid in combination are also recommended [13,15]. The organism infecting our patient in the present case was found to be resistant to ticarcillin-clavulanic acid; hence he was treated with a combination of levofloxacin and trimethoprim-sulfamethoxazole, resulting in a successful clinical outcome. However, it is to be noted that, resistance to trimethoprim-sulfamethoxazole and levofloxacin have been found in 26 % and 24 % of Indian isolates, respectively [20]. Earlier, the results from the SENTRY Antimicrobial Surveillance Program in 2004, had showed a level of resistance to trimethoprim-sulfamethoxazole of 3.8 % for *S. maltophilia* isolates [21].

Conclusion

The present case highlights the occurrence of pyogenic liver abscess due to a hitherto relatively uncommon etiologic agent, *S. maltophilia*. Owing to its increasing occurrence as an opportunistic pathogen combined with a multidrug resistant nature, its emerging role in various other pyogenic infections needs to be re-defined. Physicians also need to be aware that *S. maltophilia* infections are not restricted to hospitalized patients and may occur as an important emerging pathogen in community-acquired infections as well. Increased awareness combined with timely diagnosis and targeted antibiotic therapy is pivotal in such scenarios to ensure a favorable outcome in the clinical course.

Author's contributions

Srujana Mohanty – provided substantial contribution to the concept and design of the study; acquisition, analysis, and interpretation of data, did the literature search and revised the work for important intellectual content. She is the Corresponding author who gave the final approval for the manuscript to be published.

Shehnaz Firdaus – provided substantial contribution to the acquisition, analysis, and interpretation of data for the work, did the literature search, and drafted the manuscript.

Rashmi Ranjan Mohanty – was the treating physician and contributed to the acquisition, analysis, and interpretation of data for the study as well as critically revised the work for important intellectual content.

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Declaration of Competing Interest

The authors report no declarations of interest.

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